

## Non-invasive sinonasal mucormycosis in a diabetic - Case report

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### ABSTRACT

The case of a diabetic patient hospitalized with a discharging ulcer sinus in the maxillary area and poor oral hygiene related to mucormycosis were shown and discussed in this study. A 52-year-old man with uncontrolled diabetes was referred to a tertiary care hospital with a two-year history of headaches, toothaches, nasal obstruction, and head heaviness. Recurrent URI and side face discomfort are also common. The patient was observed with ulcer sinus drainage and left maxillary pain. In the upper second and third molar regions, there is a blackish discoloration. A histological examination showed fungal hyphae after he had surgical debridement. Amphotericin B was used as a systemic antifungal. Sinonasal mucormycosis is a disease that is currently poorly understood and has a significant fatality rate. Currently, the trifecta of physician awareness, rapid initiation of therapy, and timely surgical intervention represents the most successful method of illness management.

**Keywords:** Mucormycosis, sinonasal, antifungal, diabetes, head ache, maxillary region and Amphotericin B

### 1. INTRODUCTION

Mucormycosis is a potentially deadly illness caused by Mucorales species of the phylum Zygomycota. It occurs predominantly in immunocompromised hosts, notably those with diabetes mellitus, leukaemia, and lymphoma (Rutar and Cockerham, 2006).

Mucormycosis has a prevalence rate of 0.14 per 1000 in the Indian population, which is nearly 80 times greater than in affluent nations. Mucormycosis has a 46 percent mortality rate worldwide (Chander et al., 2018). Factors such as intracranial or orbital involvement, as well as permanent immune suppression, can elevate mortality rates to as high as 50% to 80%. In diabetic and immunocompromised individuals, a strong suspicion for this condition must be evaluated. Tissue necrosis, a defining feature of mucormycosis, is frequently a late symptom (Maini et al., 2021).

Immunocompromised individuals, such as those with poorly managed diabetes, blood dyscrasias, malnutrition, neutropenia, iron overload, organ transplantation, and immunosuppressive medication, are frequently affected.

Histopathological proof of the organism in the afflicted tissue confirms the diagnosis (Oladeji et al., 2013). Because of the disease's aggressive nature, early identification and treatment of mucormycosis are critical. Antifungal therapy should be supplemented with surgical debridement of all necrotic tissues to establish control of the underlying illness, correct metabolic abnormalities, and correct metabolic abnormalities. The purpose of this case report is to present a patient with Sinonasal mucormycosis in order to raise awareness of the disease's presence in our surroundings and underline the need of maintaining a high index of suspicion.

## 2. CASE PRESENTATION

On March 29, 2022, a 52-year-old man was admitted to Out Patient Department, Sree Balaji Medical College and Hospital, a tertiary care institution, with a two-year history of headaches, toothaches, nasal blockage, and head heaviness. He had ulcer sinus leakage and left maxillary discomfort when he came in. Blackish discoloration can also be detected in the upper second and third molar areas. There was no history of hypertension, bronchial asthma, tuberculosis, or any other debilitating illnesses in the patient, nor was there any relevant family history. On admission, the deranged investigations were: Neutrophil count 51.3% (40-80%), Lymphocyte count 31.6% (22-40%), Eosinophil count 8.5% (1-6%), ESR one hour 24mm (<19mm) Fasting blood sugar (FBS) 400 mg/dL (70-110 mg/dL), Post-prandial blood sugar (PPBS) 566 mg/dL (110-140 mg/dL), HbA1c 13.3% (<6%).

Swelling and discomfort in the left eye were the patient's complaints. For the same reason, he was sent to the OPD. There was infraorbital ulceration on clinical examination. With partial ophthalmoplegia, visual acuity was 6/6, and there was no nasal discharge. For ulceration, pus and swab samples were collected and sent to microbiology department for further evaluation.

### Management and Follow up

A preliminary lateral topogram of the brain was performed. Volume scans were then performed with OM line as reference.

On the infratentorial region, Cerebellar hemispheres, 4<sup>th</sup> ventricle and Brain stem were normal. On supratentorial region, Attenuation values of brain parenchyma grey and white matter differentiations were normal. Also basal cisterns, third and lateral ventricles were normal.

Surgical debridement was performed under general anaesthesia mostly for biopsy specimens. Under General Anaesthesia, using 0 degree endoscope crusts removed in left nasal cavity. Left middle meatal antrostomy and medial maxillectomy was done and tissue taken sent for biopsy. Debridement and Amphotericin B (1 mg/mL) lavage were performed. 6-0 resorbable suture was used to close the wound.

Three biopsy specimens were collected from the patient,

Specimen A- biopsy from left maxillary cavity

Specimen B- biopsy from left cheek ulcerative wound

Specimen C- biopsy from left maxillary cavity and nasal cavity. [Received multiple bony tissue fragments measuring 2 cm]. AE in 2 blocks [C1 and C2]

Specimen A and B: Section studied show blood clot, acute and chronic inflammatory cell infiltrates, a tiny strip of respiratory epithelium and few islands of necrosis. Occasional multinucleated giant cells are also seen.

Specimen C1: Section studied shows large area of necrosis, islands of bone, blood clot and a ball of fungal hyphae –Broad, irregular and aseptate. Few areas of inflammatory granulation tissue also seen.

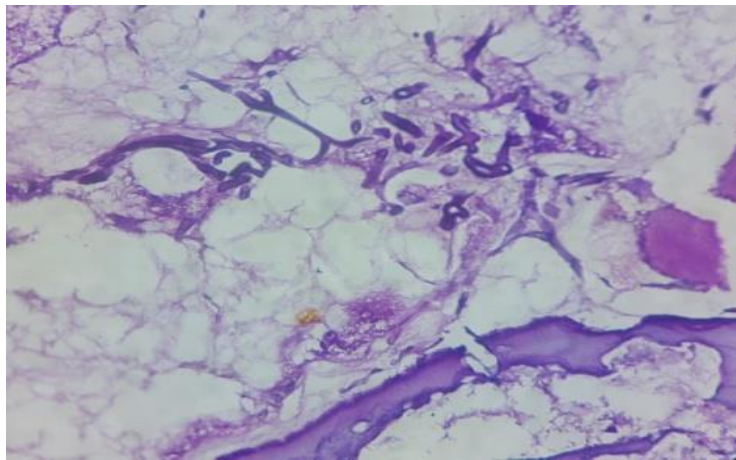
Specimen C2: Section studied shows large areas of spicules of necrotic bone and inflammatory granulation tissue with necrosis.

The levels of FBS and PPBS fell to 200 mg/dL and 284 mg/dL, respectively. The patient was pleased with the therapy and result.

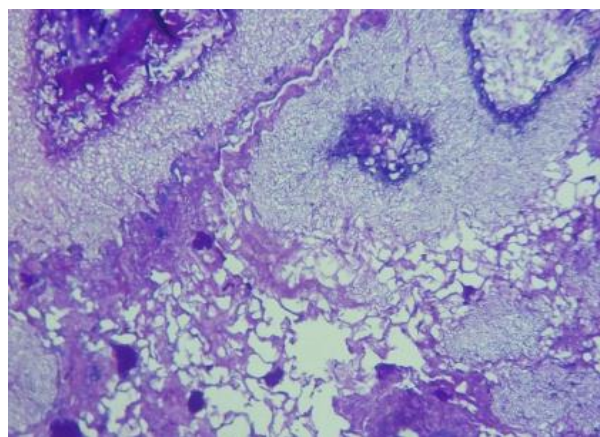
### Microbiology

Tissue biopsies were inoculated on Sabouraud's agar and Blood agar, incubated at 30 and 37 degrees Celsius, and the sample was inspected microscopically. After staining with lactofuchsin, fungal growth was examined macroscopically at 37°C and 45°C. Mucormycosis colonies were seen on the medium after 4 days. PAS (Figure 1), Garland (Figure 2) and GMS (Figure 3) were positive for fungal hyphae in special stains.

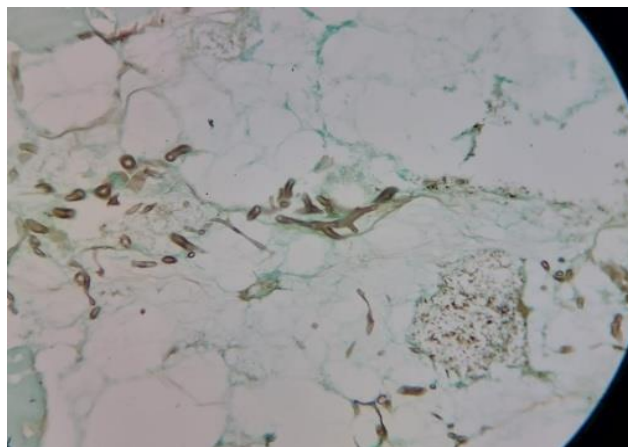
There were aseptate, branching broad-based fungal hyphae, necrosis, epithelioid cell granulomas with epithelioid cells, multinucleated giant cells, and chronic inflammatory cell infiltration. Bacterial growth was seen on blood agar, which was later identified as *E.coli* and *Klebsiella oxytoca* following biochemical analysis.



**Figure 1** PAS Staining



**Figure 2** Garland staining



**Figure 3** GMS staining

### 3. DISCUSSION

Anatomic site localization, rather than mycologic categorization, is used to suggest Mucormycosis nomenclature. Mucormycosis of the head and neck can be characterized as isolated nasal, rhino-orbital, or rhino-orbital-cerebral. Pulmonary, disseminated, cutaneous, gastrointestinal, and other types are also acceptable (Safi et al., 2020). The majority of clinical isolates are fungi from the genus *Rhizopus* (Chowdhary et al., 2014). Mucoraceae are saprophytic fungi that live on decaying debris and may also be found in bread, soil, air, dust, and hospital wards. The usage of air conditioners might conceivably be linked to seasonal variation. In temperate regions, the organisms are quite effective (Sarfaty et al., 2020). Diabetes, immunosuppressive medication, leukemias, and

neutropenias are the most prevalent risk factors. Neutrophil malfunction, hematopoietic stem cell transplantation, diabetic ketoacidosis, iron overload, and HIV/AIDS patients are among the patients (Petrikos and Drogari-Apiranthitou, 2011).

The mold normally enters the body through the lungs and has a strong affinity for arteries, growing along the internal elastic lamina, producing thrombosis and infarction. The illness progresses either directly or indirectly by vascular blockage from the nose and sinuses. Invasion through the superior orbital fissure, ophthalmic arteries, cribriform plate, carotid artery, or potentially a perineural pathway can also cause intracranial involvement (Ghagane and Nerli, 2021). Previous research has shown that diabetic ketoacidosis reduces neutrophil chemotaxis and phagocytic activity while also increasing accessible serum iron (Morales-Franco et al., 2021). The prognosis is influenced by a number of factors, one of which is the timing of therapy. Once the diagnosis is established, the patient is started on conservative treatment. Due to worries about impairment and deformity, orbital exenteration remains the most difficult option in rhino-orbital instances. Although exenteration is the last option, it can save a person's life at the cost of mutilation.

During the early stages of rhinocerebral mucormycosis, imaging modalities are of little use, with thickening of the sinus mucosa or extraocular muscles being mentioned as an early symptom of the disease. CT scans can be used to assess disease progression, albeit connection with clinical findings is not always reliable (Mazzai et al., 2022). Although amphotericin B deoxycholate (AMP) is still the only antifungal treatment approved for mucormycosis, lipid formulations of amphotericin B offer a safe and effective alternative. For the lipid formulation of amphotericin, initial doses of 5-10 mg/kg/day are indicated, with higher doses (up to 10 mg/kg/day) recommended for CNS involvement (Mallis et al., 2010).

#### 4. CONCLUSION

Sinonasal mucormycosis is a disease that is currently poorly understood and has a significant fatality rate. Currently, the trifecta of physician awareness, rapid therapy start, and timely surgical intervention represents the most successful method of illness management. The treatment of mucormycosis patients should be multidisciplinary and started as soon as possible. Patients should be given amphotericin B as soon as possible after surgery. There is no agreement on the overall therapy time, which varies depending on the clinical progress of each patient.

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#### Conflicts of interest

The authors declare that there are no conflicts of interests.

#### Data and materials availability

All data associated with this study are present in the paper.

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